Research Article

Decrease of spasticity after hybrid assistive limb® training for a patient with C4 quadriplegia due to chronic SCI

Akira Ikumi[1], Shigeki Kubota[1,2], Yukiyo Shimizu[3], Hideki Kadone[4], Aiki Marushima[5], Tomoyuki Ueno[3], Hiroaki Kawamoto[6], Yasushi Hada[3], Akira Matsumura[5], Yoshiyuki Sankai[6], Masashi Yamazaki[1,4]

[1]Department of Orthopaedic Surgery, Faculty of Medicine, University of Tsukuba, Ibaraki, Japan, [2]Division of Regenerative Medicine for Musculoskeletal System, Faculty of Medicine, University of Tsukuba, Ibaraki, Japan, [3]Department of Rehabilitation Medicine, University of Tsukuba Hospital, Ibaraki, Japan, [4]Center for Innovating Medicine and Engineering (CIME), University of Tsukuba Hospital, Ibaraki, Japan, [5]Department of Neurosurgery, Faculty of Medicine, University of Tsukuba, Ibaraki, Japan, [6]Faculty of Systems and Information Engineering, University of Tsukuba, Ibaraki, Japan

Context: Recently, locomotor training with robotic assistance has been found effective in treating spinal cord injury (SCI). Our case report examined locomotor training using the robotic suit hybrid assistive limb (HAL) in a patient with complete C4 quadriplegia due to chronic SCI. This is the first report examining HAL in complete C4 quadriplegia.

Findings: The patient was a 19-year-old man who dislocated C3/4 during judo 4 years previously. Following the injury, he underwent C3/4 posterior spinal fusion but remained paralyzed despite rehabilitation. There was muscle atrophy under C5 level and no sensation around the anus, but partial sensation of pressure remained in the limbs. The American Spinal Injury Association impairment scale was Grade A (complete motor C4 lesion).

HAL training was administered in 10 sessions (twice per week). The training sessions consisted of treadmill walking with HAL. For safety, 2 physicians and 1 therapist supported the subject for balance and weight-bearing. The device's cybernic autonomous control mode provides autonomic physical support based on predefined walking patterns.

We evaluated the adverse events, walking time and distance, and the difference in muscle spasticity before and after HAL-training using a modified Ashworth scale (mAs).

No adverse events were observed that required discontinuation of rehabilitation. Walking distance and time increased from 25.2 meters/7.6 minutes to 148.3 meter/15 minutes. The mAs score decreased after HAL training.

Conclusion: Our case report indicates that HAL training is feasible and effective for complete C4 quadriplegia in chronic SCI.

Keywords: Hybrid assistive limb (HAL), Spinal cord injury, Locomotor training, Robotics, Spasticity

Introduction

Rehabilitation robotics emerged in the 1980s with the aim of using robotic technology to assist people with movement dysfunction.[1] Robotic devices have recently been developed for use in clinical settings.

Tefertiller *et al*.[2] reviewed 30 articles (14 randomized controlled trials, 16 nonrandomized controlled trials) that examined the effects of locomotor training with robotic assistance in patients after stroke, spinal cord injury (SCI), multiple sclerosis, traumatic brain injury, and Parkinson's disease. The review supports the conclusion that locomotor training with robotic assistance is beneficial for improving walking function in individuals after stroke and SCI.[2]

The development of main gait training machines followed. These machines either involve an exoskeleton robotic device (e.g. Lokomat®, LOPES exoskeleton

Correspondence to: Akira Ikumi, Department of Orthopaedic Surgery, Faculty of Medicine, University of Tsukuba, 1-1-1, Tennodai, Tsukuba, Ibaraki, 305-8575, Japan. Email: ikumi@tsukuba-seikei.jp

DOI 10.1080/10790268.2016.1225913

robot)[3,4] or a robotic device with foot-driven plates (e.g. Gait Trainer GT I®, Haptic Walker).[5,6] The exoskeleton robotic device is equipped with programmable drives or passive elements that flex the knees and hips during the swing phase, whereas with the other type of robotic device, the feet are placed on footplates whose trajectories simulate the stance and swing phases.

Other than robotic gait training and conventional therapy, another treatment approach involves treadmill training with partial body weight support.[7] However, this approach requires considerable involvement of a physical therapist, and generally, 3 therapists are required to induce movement of the paretic leg during the swing phase and to shift the patient's weight onto the stance limb.

The potentially positive common benefits of robotic gait training are that it involves repeatedly undergoing sufficient and accurate training for a prolonged period. Lokomat is the first robotic-driven gait orthosis with electromechanical drives to assist the walking movements of gait-impaired patients on a treadmill by supporting the body weight.[8,9] Husemann *et al.*[10] compared a Lokomat group that received 30 minutes of robotic training with a control group that received 30 minutes of conventional physiotherapy. After 4 weeks of therapy, although there was no significant difference in walking ability between the groups, the walking ability in both groups as expressed by functional ambulation classification was significantly improved. The researchers reported that the Lokomat group demonstrated an advantage for robotic training over conventional physiotherapy in the improvement of gait abnormality and body tissue composition.[10]

However, in a recent randomized controlled study[11] that compared robot-assisted locomotor training with therapist-assisted locomotor training in chronic stroke patients, the results indicated that greater improvements in speed and single limb stance time on the impaired leg were observed in subjects who received therapist-assisted locomotor training. Thus, the usefulness of robot-assisted rehabilitation is controversial.

The hybrid assistive limb® (HAL®; Cyberdyne Inc, Ibaraki, Japan)[12–15] is a wearable robotic suit that assists in voluntary control of knee- and hip-joint motion (Fig. 1). Signals from force-pressure sensors in the shoes and muscle action potentials detected through electrodes on the surface of the skin are processed through a computer, and assisted motions are provided to the patient. Power units on the hip and knee joints on both sides consist of angular sensors and actuators, and the control system consists of a cybernic voluntary control (CVC) and a cybernic autonomous control (CAC) subsystem.[12]

HAL has been reported to be useful in the functional recovery of various mobility disorders.[12,16–18] To the best of our knowledge, however, there is no published report to clarify the feasibility of rehabilitation with HAL for a patient with complete quadriplegia. Therefore, the efficacy and safety of HAL for complete quadriplegia remains unclear.

In the current case report, HAL training was performed for a patient with complete quadriplegia after SCI, and efficiency and safety were evaluated. This study was conducted with the approval of the Ethics Committee of the Tsukuba University Faculty of Medicine.

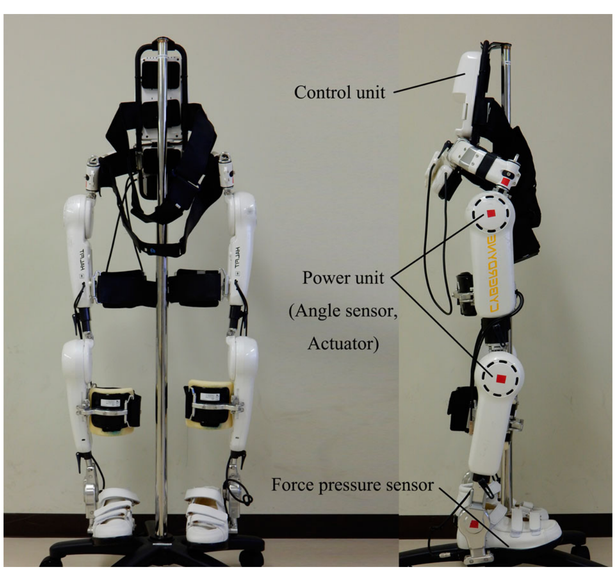

Figure 1. The robotic suit HAL. (Colour online)

Case presentation

Patient

A 19-year-old man who was injured while participating in judo 4 years previously was diagnosed with a cervical vertebral fracture-dislocation (C3/4). Emergency surgery (posterior spinal fusion) was performed. After surgery, the patient required respiratory care with a ventilator, but at one month postoperatively, he no longer required the ventilator. Complete paralysis and serious sensory dysfunction inferior to the C5 level were present from the time of injury. He continued the rehabilitation during the hospital stay and ambulatory rehabilitation after discharge. In spite of the aggressive rehabilitation, his paralysis had scarcely improved, and he required assistance with all activities of daily living. He was hospitalized in our facility to undergo rehabilitation using the robotic suit HAL.

Physical examination findings on admission indicated that the patient required comprehensive care, including feeding, changing clothes, bathing and egestion. He used the chin-controlled electric wheelchair to move outside. The neurologic examination revealed muscle weakness with a manual muscle testing (MMT) score of 5/5 in the trapezius muscle and an MMT score of 0/0 below the deltoid muscle (C5 level). The patient had severe sensory disturbances below the C5 level. A slight sense of pressure remained in his right upper extremity and both lower extremities, but there was none in other areas. No articular contracture was observed. No urinary bladder or bowel function remained. The results of the blood and urine tests were normal.

The computed tomography (CT) and the magnetic resonance imaging (MRI) immediately after injury showed a C3/4 vertebral fracture-dislocation and interlocking of the right facets. The spinal cord was compressed tightly (Fig. 2). The radiographic findings after surgery showed posterior spinal fusion between C3 and C4. The vertebral dislocation was reduced well (Fig. 3). The CT and MRI findings on admission showed no loosening of the implant and decompression at the injury site of the spinal cord. A signal change (low signal at T1WI and high signal at T2WI) of the spinal cord was observed (Fig. 4).

Clinical evaluation before HAL training showed the following: the American Spinal Injury Association (ASIA) impairment scale (AIS) was grade A (complete motor C4 lesion); the ASIA motor score (lower limb total) was 0 points; the ASIA sensory score for light touch was 62 points (right: 31 points; left: 31 points); the Frankel classification was grade B2; the Spasm Frequency Score was 3 (spasm occurred 1 to 10 times per hour); the Barthel Index was 5/100 points; the Total Functional Independence Measure Score was 53/126 points (motor 18/91 points, cognitive 35/35 points); and the Functional Balance Scale was 0/56 points.

HAL training

The patient received additional HAL training 2 times per week for 5 weeks (10 sessions) in addition to standard physical and occupational therapy. HAL training lasted 60 minutes, including rests and time for attaching/detaching the device. At the initiation of HAL training, the robot was fitted, and the sitting/standing motion was confirmed. The training sessions consisted of treadmill walking with HAL. A body weight support system (945-480 Unweighing System, BIODEX®, Shirley, NY, USA) with a harness was used for safety.

The cybernic autonomous control (CAC) mode provides autonomic physical support based on predefined walking patterns from able-bodied persons. For safety reasons, 2 physicians and 1 therapist supported the subject in balance and weight bearing (Fig. 5).

We evaluated the walking time and distance, the modified Ashworth scale score (mAs)[19] before and after HAL training, and adverse events associated with HAL training.

The time from attaching the device to setting the unweighing system was an average of 10 minutes. Walking distance and time increased from 25.2 meters/7.6 minutes (first session) to 148.3 meters/7.6 minutes (last session) (Figs. 6 and 7). The total mAs score (Score: 0–144; the number of joints: 36) was evaluated before and after HAL training. The score before HAL training was 15.13 ± 2.80 points; after HAL training, it was 5.75 ± 2.38 points. No joint change for the worse after training was observed (Fig. 8). The average number of joints decreased, and the spasticity was 7. The efficiency continued for approximately 30 minutes after HAL training. There were no adverse events requiring discontinuation of the HAL training. A transient blood pressure change (systolic blood pressure <90 or >180) was observed 6 times/10 sessions (0.6 times/session), but the blood pressure returned to baseline after a few minutes of resting.

Discussion

Aach *et al.*[18] demonstrated the clinical potential of HAL training based on voluntary drive in patients suffering from chronic SCI. Fujii *et al.*[20] reported that the training using an advanced robotic device may affect the patient's motivation for rehabilitation based on the analysis of questionnaires from patients undergoing HAL training.

On the other hand, Maeshima *et al.*[16] reported that the HAL suit should not be used in a patient with paralysis severe enough to cause muscle contraction or whose bioelectric signals cannot be sensed. Thus, the use of HAL for patients with severe chronic SCI is still controversial.

In this case, we investigated the feasibility of rehabilitation using a robotic suit HAL for C4 quadriplegia, and confirmed that HAL training could be implemented safely. No serious HAL training-related adverse events occurred. Furthermore, the walking time and distance had increased as the rehabilitation continued, suggesting the learning effect of the HAL training for the patient with complete C4 quadriplegia.

In our case, a certain effect on decreasing the spasticity was also confirmed after HAL training. Spasticity

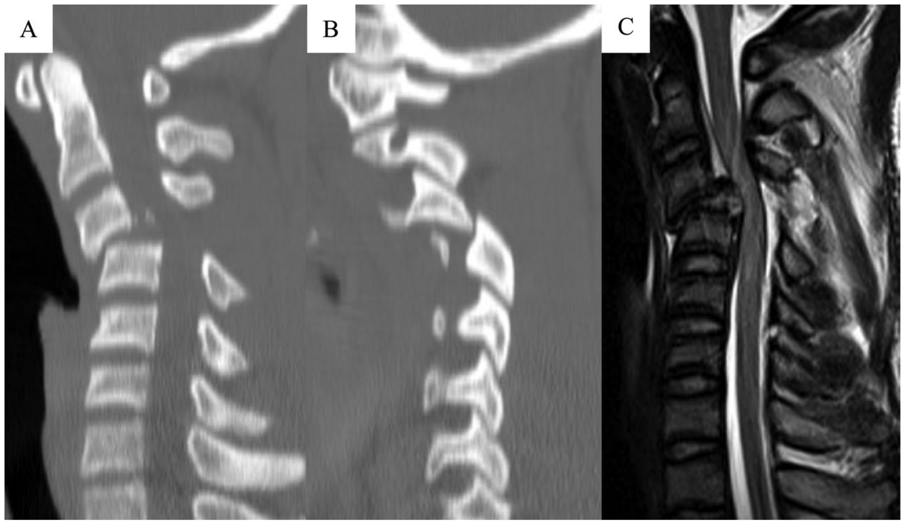

Figure 2. Mid-sagittal (A) and right-lateral (B) reconstruction images of CT and a mid-sagittal view of T2-weighted MR image immediately after injury. The CT images show the cervical vertebral fracture-dislocation at C3/4 (A, B). The MR image shows the compressed spinal cord at C3/4 (C).

was defined as “a motor disorder characterized by velocity-dependent increase in tonic stretch reflexes (muscle tone) with exaggerated tendon jerks, resulting from hyper-excitability of stretch reflexes, as a main component of upper motoneuron syndrome.[21] The spasticity after SCI is a serious hindrance factor at the start of active rehabilitation.[22] The decrease of spasticity is directly linked to the functional improvement of the SCI patient. Powell *et al.* [23] reported the usefulness of combined therapy in transvertebral direct current stimulation (tvDCS) and locomotor training on a robot-assisted gait orthosis (LT-RGO) for spasticity, and Duffel *et al.*[24] reported that robotic locomotor training with anti-spastic medication improves the walking ability by decreasing spasticity. On the other hand, a systematic review found that the effects of robot-assisted therapy on muscle spasticity were inconsistent.[25] Several studies have reported that prolonged passive muscle stretching reduces spasticity.[26–28] Sustained ambulation activity due to HAL training have possibility to effect similar decreasing of spasticity as

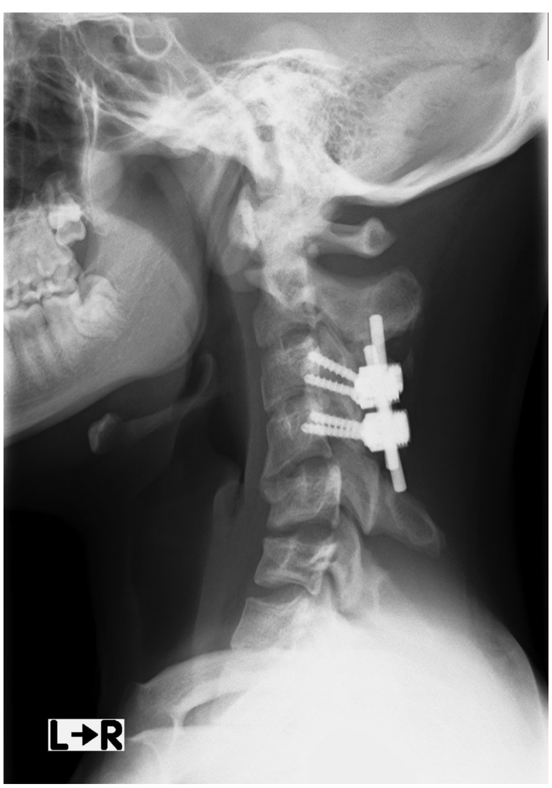

Figure 3. A cervical lateral radiograph 4 years after surgery, showing the completion of C3/4 posterior spinal fusion.

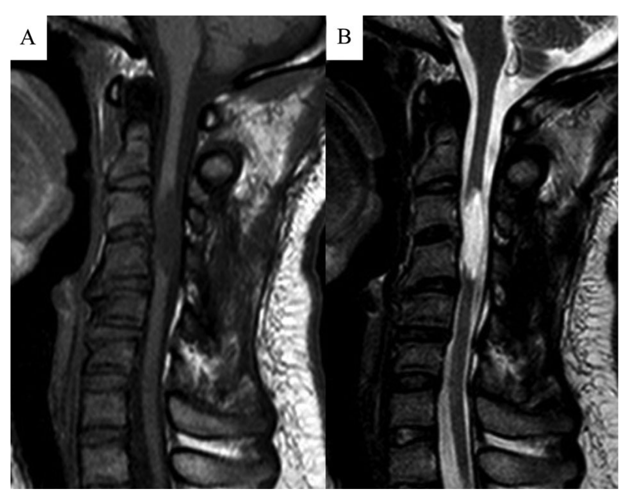

Figure 4. Mid-sagittal T1-weighted (A) and T2-weighted (B) MR images 4 years after surgery. The MR images show the signal changes in the spinal cord at the C3/4 level.

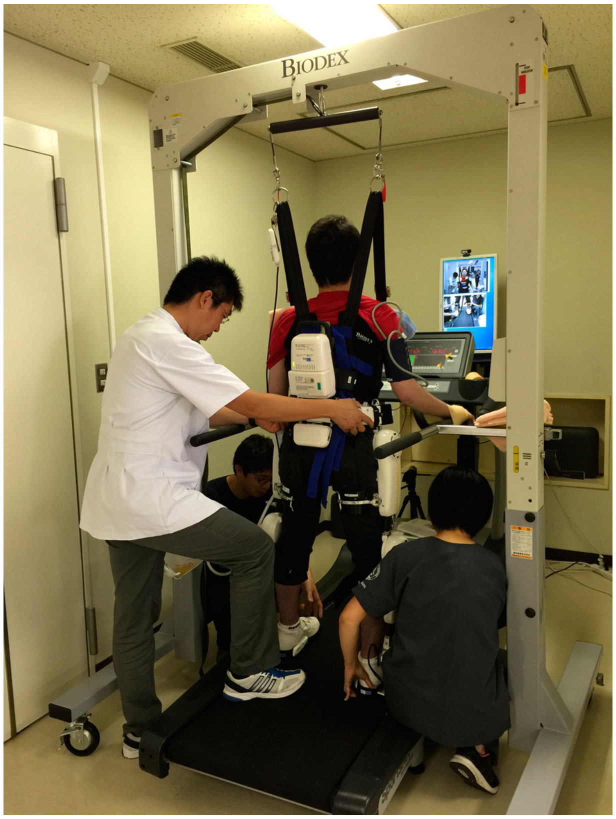

Figure 5. Gait training using the robotic suit HAL with a body weight support system (BIODEX). (Colour online)
2 physicians and 1 therapist support the subject in balance and weight-bearing.

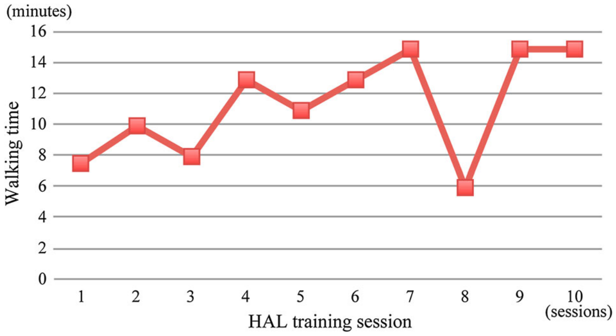

Figure 6. Walking time in each HAL session.

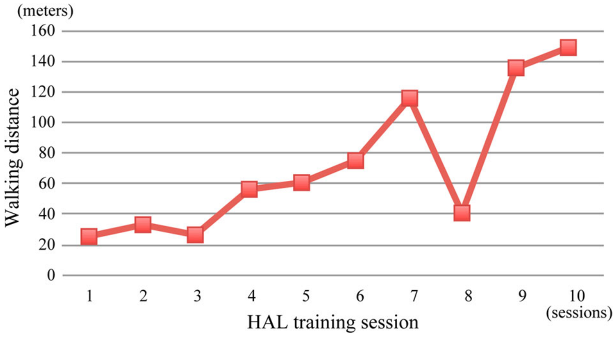

Figure 7. Walking distance in each HAL session.

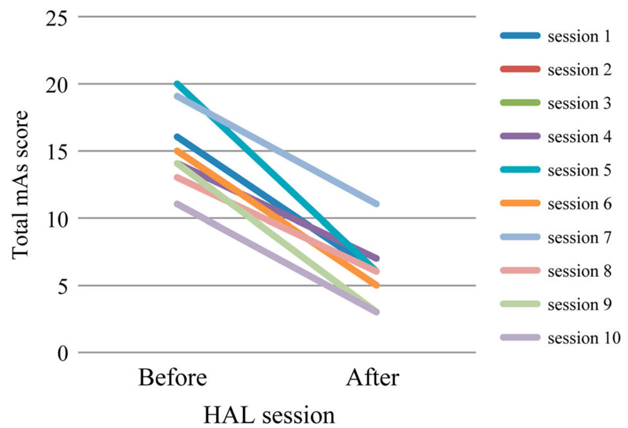

Figure 8. Change in the total modified Ashworth scale score before and after training in each HAL session. (Colour online)

passive muscle stretching. More studies are required in order to verify the availability of HAL training. To the best of our knowledge, however, there has been no report that a single robotic locomotor training decreased the spasticity of patients with SCI. In this meaning, this is the first report that the HAL training has a possibility to decrease the spasticity of patients with SCI, in spite of the fact that the decrease of spasticity in our patient was temporary (lasting approximately 30 minutes after HAL training).

In summary, the HAL training for a patient with complete C4 quadriplegia and chronic SCI decreased the spasticity, indicating the feasibility and efficiency of rehabilitation using a robotic suit HAL for quadriplegia patients.

Acknowledgements

We thank Mayuko Sakamaki and Yumiko Ito, Center for Innovative Medicine and Engineering (CIME), University of Tsukuba Hospital, for their excellent technical assistance. This study was supported by the Industrial Disease Clinical Research Grants of the Ministry of Health Labour and Welfare, Japan (14060101-01).

Disclaimer statements

Contributors None.

Funding None.

Conflicts of interest None.

Ethics approval None.

ORCiD

Akira Ikumi http://orcid.org/0000-0001-6034-1593

References

1 van Vliet P, Wing AM. A new challenge: robotics in the rehabilitation of the neurologically motor impaired. Phys Ther 1991;71(1):39–47.
2 Tefertiller C, Pharo B, Evans N, Winchester P. Efficacy of rehabilitation robotics for walking training in neurological disorders: a review. J Rehabil Res Dev 2011;48(4):387–416.
3 Colombo G, Joerg M, Schreier R, Dietz V. Treadmill training of paraplegic patients using a robotic orthosis. J Rehabil Res Dev 2000;37(6):693–700.
4 Veneman JF, Kruidhof R, Hekman EE, Ekkelenkamp R, Van Asseldonk EH, van der Kooij H. Design and evaluation of the LOPES exoskeleton robot for interactive gait rehabilitation. IEEE Trans Neural Syst Rehabil Eng 2007;15(3):379–86.
5 Hesse S, Uhlenbrock D, Werner C, Bardeleben A. A mechanized gait trainer for restoring gait in nonambulatory subjects. Arch Phys Med Rehabil 2000;81(9):1158–61.
6 Schmidt H, Werner C, Bernhardt R, Hesse S, Kruger J. Gait rehabilitation machines based on programmable footplates. J Neuroeng Rehabil 2007;4:2.
7 Visintin M, Barbeau H, Korner-Bitensky N, Mayo NE. A new approach to retrain gait in stroke patients through body weight support and treadmill stimulation. Stroke 1998;29(6):1122–8.
8 Lunenburger L, Colombo G, Riener R, Dietz V. Biofeedback in gait training with the robotic orthosis Lokomat. Conf Proc IEEE Eng Med Biol Soc 2004;7:4888–91.
9 Neckel N, Wisman W, Hidler J. Limb alignment and kinematics inside a Lokomat robotic orthosis. Conf Proc IEEE Eng Med Biol Soc 2006;1:2698–701.
10 Husemann B, Muller F, Krewer C, Heller S, Koenig E. Effects of locomotion training with assistance of a robot-driven gait orthosis in hemiparetic patients after stroke: a randomized controlled pilot study. Stroke 2007;38(2):349–54.
11 Hornby TG, Campbell DD, Kahn JH, Demott T, Moore JL, Roth HR. Enhanced gait-related improvements after therapist- versus robotic- assisted locomotor training in subjects with chronic stroke: a randomized controlled study. Stroke 2008;39(6):1786–92.
12 Kawamoto H, Sankai Y. Power assist method based on phase sequence and muscle force condition for HAL. Adv Robot 2005;19:717–34.
13 Lee S, Sankai Y. Virtual impedance adjustment in unconstrained motion for an exoskeletal robot assisting the lower limb. Adv Robot 2005;19:773–95.
14 Suzuki K, Gouji M, Kawamoto H, Hasegawa Y, Sankai Y. Intention- based walking support for paraplegia patients with robot suit HAL. Adv Robot 2007;21:1441–69.
15 Tsukahara A, Kawanishi R, Hasegawa Y, Sankai Y. Sit-to-stand and stand-to-sit transfer support for complete paraplegic patients with robot suit HAL. Adv Robot 2010;24:1615–38.
16 Maeshima S, Osawa A, Nishio D, Hirano Y, Takeda K, Kigawa H, *et al*. Efficacy of a hybrid assistive limb in post-stroke hemiplegic patients: a preliminary report. BMC Neurol. 2011;11:116.
17 Kubota S, Nakata Y, Eguchi K, Kawamoto H, Kamibayashi K, Sakane M, *et al*. Feasibility of rehabilitation training with a newly developed wearable robot for patients with limited mobility. Arch Phys Med Rehabil 2013;94(6):1080–7.
18 Aach M, Cruciger O, Sczesny-Kaiser M, Hoeffken O, Meindl R, Tegenthoff M, *et al*. Voluntary driven exoskeleton as a new tool for rehabilitation in chronic spinal cord injury: a pilot study. Spine J. 2014;14(12):2847–53.
19 Bohannon RW, Smith MB. Inter-rater reliability of a modified Ashworth scale of muscle spasticity. Phys Ther 1987;67:206–7.
20 Fujii K, Abe T, Kubota S, Marushima A, Kawamoto H, Ueno T, *et al.* The voluntary driven exoskeleton Hybrid Assistive Limb (HAL) for postoperative training of thoracic ossification of the posterior longitudinal ligament: a case report. J Spinal Cord Med 2016;Feb 9. [Epub ahead of print]
21 Lance JW. What is spasticity? Lancet 1990;335(8689):606.
22 van Cooten IP, Snoek GJ, Nene AV, de Groot S, Post MW. Functional hindrance due to spasticity in individuals with spinal cord injury during inpatient rehabilitation and 1 year thereafter. Spinal Cord 2015;53(9):663–7.
23 Powell ES, Carrico C, Raithatha R, Salyers E, Ward A, Sawaki L. Transvertebral direct current stimulation paired with locomotor training in chronic spinal cord injury: a case study. NeuroRehabilitation 2016;38(1):27–35.
24 Duffell LD, Brown GL, Mirbagheri MM. Facilitatory effects of anti-spastic medication on robotic locomotor training in people with chronic incomplete spinal cord injury. J Neuroeng Rehabil 2015;12:29.
25 Peter O, Fazekas G, Zsiga K, Denes Z. Robot-mediated upper limb physiotherapy: review and recommendations for future clinical trials. Int J Rehabil Res 2011;34(3):196–202.
26 Pin T, Dyke P, Chan M. The effectiveness of passive stretching in children with cerebral palsy. Dev Med Child Neurol 2006;48(10):855–62.
27 Stevenson VL. Rehabilitation in practice: spasticity management. Clin Rehabil 2010;24(4):293–304.
28 Tremblay F, Malouin F, Richards CL, Dumas F. Effects of prolonged muscle stretch on reflex and voluntary muscle activations in children with spastic cerebral palsy. Scand J Rehabil Med 1990;22(4):171–80.